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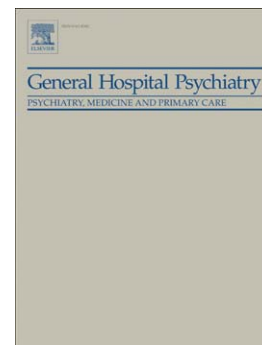
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# **Factitious Disorder: a systematic review of 455 cases in the professional literature**

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## ABSTRACT

**Objective:** Patients with factitious disorder (FD) fabricate illness, injury or impairment for psychological reasons and, as a result, misapply medical resources. The demographic and clinical profile of these patients has yet to be described in a sufficiently large sample, which has prevented clinicians from adopting an evidence-based approach to FD. The present study aimed to address this issue through a systematic review of cases reported in the professional literature.

**Method:** A systematic search for case studies in the MEDLINE, Web Of Science, and EMBASE databases was conducted. 4092 records were screened and 684 remaining papers reviewed. A supplementary search was conducted via Google Scholar, reference lists of eligible articles, and key review papers. In total, 372 eligible studies yielded a sample of 455 cases. Information extracted included: age, gender, reported occupation, comorbid psychopathology, presenting signs and symptoms, severity, and factors leading to the diagnosis of FD.

**Results:** 66.2% of patients in our sample were female. Mean age at presentation was 34.2 years. A healthcare or laboratory profession was reported most frequently (N=122). A current or past diagnosis of depression was described more frequently than personality disorder in cases reporting psychiatric comorbidity (41.8% versus 16.5%) and more patients elected to self-induce illness or injury (58.7%) than simulate or falsely report it. Patients were most likely to present with endocrinological, cardiological and dermatological problems. Differences among specialties were observed on demographic factors, severity, and factors leading to diagnosis of FD.

**Conclusions:** Based on the largest sample of patients with FD analysed to date, our findings offer an important first step towards an evidence-based approach to the disorder. Future guidelines must be sensitive to differing methods used by specialists when diagnosing FD.

**Key words:** Factitious disorder, Munchausen syndrome, medical deception, fabricated illness, medically unexplained symptoms, psychosomatic medicine.

## INTRODUCTION

Factitious disorder with physical symptoms (FD) is a psychiatric disorder in which sufferers intentionally fabricate illness, injury, or impairment in order to gain hospital admission and undergo medical procedures, without any obvious gain (American Psychiatric Association, 2013). It is considered to be one of the most challenging disorders in medical experience (Feldman and Eisendrath, 1996). Patients with FD may exaggerate or lie about a medical condition, mimic or “act out” medical symptoms, interfere with diagnostic investigations, or even directly self-induce illness or injury (Feldman, 2008). In contrast to malingers, who fabricate medical need for reasons of clear external reward (such as evading military service or gaining disability benefits), the motivations of patients with FD are ‘almost always obscure’ (World Health Organisation, 1992) and may include: a desire to receive affection and care, an “adrenaline rush” from undergoing medical procedures, or a sense of control from deceiving healthcare professionals (Lawlor and Kirakowski, 2014). Patients with FD may expose themselves to a considerable risk of iatrogenic harm (DeWitt et al., 2009). Indeed, one patient with FD described by Robertson and Hossain (Robertson and Hossain, 1997) admitted to having undergone 42 surgical procedures over the course of 850 admissions to 650 different hospitals. Fatality due to FD appears to be rare, but does occur (Vadugathan et al., 2014; Hirayama et al., 2003; McEwen, 1998; Nichols et al., 1990).

Studies of FD demonstrate the heavy impact of unnecessary investigations, treatments, and hospital admissions on the healthcare system. Healthcare costs in individual cases of FD have exceeded \$200,000 (Romano et al., 2014) and even \$1,000,000 (Bright et al., 2001).. A patient with FD may also have a considerable psychological impact on hospital staff involved in their care. Staff may feel anger at having been “duped” by the patient and “cheated” of the time and support they have expended (Crawford et al., 2005), or they may experience guilt for allowing themselves to be drawn into the emotional conflicts that frequently arise in cases of FD (Chambers et al., 2007; Stiles et al., 2001).

Most doctors will encounter at least one patient with FD over the course of their clinical practice (Ogbonmwan and Abidogun, 2005). However, the exact prevalence of FD in hospital settings is currently unknown (Kahn et al., 2015; Oner et al., 2015; Patenaude et al., 2006). FD may account for between 0.6 and 3% of referrals from general medicine to psychiatry (Fliege et al., 2002; Kapfhammer, 1998; Sutherland and Rodin, 1990) and between 0.02 and 0.9% of cases reviewed in specialist clinics (Mailis-Gagnon et al., 2008; Bhargava et al., 2007; Ballas, 1996; Bauer, 1996). A recent study surveying physicians' own estimates of the presence of factitious symptoms among their patients reported a higher prevalence rate of 1.3% (Fliege et al., 2007). Rates of FD may be greatly increased in patient populations whose reported problems are diagnostically challenging (Aduan et al., 1979; Rumans et al., 1978) or have received significant public attention (Mehta and Khan, 2002).

Although FD has been recognised by clinicians for centuries (Steel, 2009), if not millenia (Gavin, 1843), the first extensive study of FD appears in Asher's initial description of "Munchausen's syndrome" in 1951. However, since that time, the term "Munchausen's syndrome" has become a source of confusion in both clinical practice and the published literature (Feldman, 2008). The correct usage of the term is to denote a particularly severe and chronic presentation of FD (Steel, 2009), but "Munchausen's" is often used interchangeably with "factitious". Other terms used for FD include "hospital hopper syndrome", "hospital hobo syndrome", and "thick chart syndrome", and they frequently display a level of irony – e.g. "black hole patients", or "peregrinating problem patients". These terms reflect that patients with FD can be derided by healthcare professionals.

Patients with FD may fabricate medical need in several ways. The variety of methods available to these patients is limited in principle only by their level of dedication, imagination, and medical knowledge (Haddad et al., 2002) but is dependent in practice upon the nature of the medical problem they intend to fabricate. For example, a patient with FD attempting to fabricate urological disease may falsely report the presence of chronic urinary discomfort, deliberately withhold urine to simulate acute

anuria (Schmidt et al., 1996), add blood to urine samples to simulate haematuria (Chew et al., 2002), or actually induce a urinary tract infection by self-injection with bacterial cultures (Savino and Fordtran, 2006). A patient attempting to fabricate a dermatological condition may be restricted to simulating a lesion (e.g. by discoloration of the skin with ink (Parent et al., 1994)) or creating an actual lesion through self-mutilation (Svirsky et al., 1987) or other means (Harper and Copeman, 1983). Patients with FD may employ several of these methods at once (Feldman, 2008) and frequently present with diverse symptomatology. The wealth of medical knowledge now available on the Internet may enable patients lacking a background in healthcare to present with complex medical problems. It is seldom possible to diagnose FD with conviction (Feldman, 2008) but when the diagnosis is made, it usually follows an exhaustive series of medical procedures undertaken to rule out an organic explanation for the patient's problems.

Early detection of FD is thus paramount in order to limit wastage of healthcare resources and harm to patients. Early management of FD may also facilitate improved outcomes for patients with the disorder (Feldman, 2008). However, the clinical and demographic profile of patients with FD has not been clarified with a sufficiently large sample (Steel, 2009). We consider such knowledge to be an important first step in the development of an evidence-based approach to the early detection and management of FD in clinical settings.. The majority of the published literature on FD consists of case reports and series, which are a valuable source of information but may in isolation present a misleading clinical picture of the disorder (Krahn et al., 2014). Indeed, assumptions about the characteristics of patients with FD abound in the professional literature – one troubling example being the idea that the majority of patients with the disorder are male, as specified in the DSM-IV despite the clear lack of research supporting such a statement (American Psychiatric Association, 1994). Although recommendations have been published concerning the detection of FD (e.g. Steel, 2009), these recommendations have not been supported by broad evidence on how FD is diagnosed by clinicians on a wider scale, or how methods for detecting medical deception may vary among medical specialties. Similarly, guidelines for management of FD (e.g. Bass and Halligan, 2014) have been written in the absence of substantial data concerning the severity of the methods typically adopted by

patients with FD – or indeed the suicide risk and psychiatric comorbidity associated with the disorder. This is information integral to effective management of FD (Stiles et al., 2001).

, What is therefore needed is a comprehensive and systematic review of the case reports and series available in the professional literature, as has been undertaken previously with child and adolescent FD (Libow, 2000), FD imposed upon another or “Munchausen-by-proxy syndrome” (Sheridan, 2003; Feldman and Brown, 2002), and other uncommon disorders (Dhir et al., 2007; Arnulf et al., 2005; Biarge et al., 2004). Use of this method has enabled authors to examine the clinical and demographic characteristics of samples of patients larger than would be feasible for comparable empirical studies.

Unfortunately, only a limited number of reviews have been published on FD, and those published to date have been mainly limited to a small number of cases from single medical specialties – recently, cardiology (Mehta and Khan, 2002), neurology (Kanaan and Wessely, 2010), obstetrics and gynecology (Edi-Osagie et al., 1998), ENT (Alicandri-Ciufelli et al., 2012), oncology (Baig et al., 2015) and dermatology (Boyd et al., 2014). Authors who have aggregated cases across specialties have limited their sample to cases of FD that have been treated (Eastwood and Bisson, 2008) or detected by laboratory testing (Kinns et al., 2013; Kenedi et al., 2011), and have therefore analysed only a minority of cases available in the professional literature.

Thus, it was the aim of this study to undertake a comprehensive, systematic review of all cases of FD with physical symptoms published in the professional literature to date, to characterise for the first time the basic demographic and clinical profile of patients with FD in a large sample, and to compare these features among medical specialties. This review was restricted to adult cases of FD, as a full review of child and adolescent FD was beyond the scope of this study and has previously been conducted (Libow, 2000).



## METHOD

### Types of study

A systematic search was conducted for all case studies and series that reported on adult patients eligible for a DSM-5 diagnosis of FD with primarily physical symptoms (American Psychiatric Association, 2013) on the basis of the clinical information provided by the author(s). This search included cases where the diagnosis of FD was described in other terms, such as ‘dermatitis artefacta’ and ‘Munchausen’s’, or classified according to a comparable diagnostic system, such as DSM-IV (American Psychiatric Association, 1994) or ICD-10 (World Health Organisation, 1992). Chart reviews and larger case series were excluded if they did not also describe cases individually. Following the conservative methodology outlined by Kanaan and Wessely (Kanaan and Wessely, 2010), studies were excluded if they reported cases in which no firm diagnosis of FD could be made.

### Search strategy

A broad keyword search of literature published in English between January 1, 1965 and July 27, 2015 was conducted. MEDLINE, Web of Science, and EMBASE databases were searched using the terms, *factit\**, *munchausen\**, *artefacta\** and *artefactua\**. Records with ‘by proxy’ or ‘imposed upon another’ were not automatically filtered out of the search results in order to ensure that case series reporting both FD and FD imposed upon another were included. 4,256 records were returned following exclusion of duplicate records, of which 4,092 were retrieved for abstract review. 748 records were identified as potentially eligible, of which 684 were retrieved for full text review. 333 studies were selected for inclusion after full text review. The bibliographies of eligible studies were also screened, in addition to the bibliographies of multiple review papers (Baig et al., 2015; Boyd et al., 2014; Kinns et al., 2013; Alicandri-Ciufelli et al., 2012; Kenedi et al., 2011; Kanaan and Wessely, 2010; Eastwood and Bisson, 2008; Edi-Osagie et al., 1998) and the results of a GoogleScholar search utilising terms

identical to the keyword search. These supplementary search processes yielded a further 39 eligible studies. Search formulae for MEDLINE, Web of Science, and EMBASE databases are provided in Section 1 of the Supplemental Material. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart for the search process is provided in Section 2 of the Supplemental Material.

### Data collected

A mean number of 9.1 new cases/year was reported over the review period, with a tendency toward higher values in more recent years: 1965-75 (3.5/year), 1975-85 (7.4/year), 1985-95 (11.2/year), 1995-2005 (12/year), and 2005-2015 (11.3/year). Single cases were extracted from 86% of studies, while the remaining 14% contributed multiple patients.

The following quantitative and qualitative variables were obtained (percentage of data found indicated in parentheses) for each case: *age* (99%), *gender* (100%), *reported occupation* (47%), *index presentation of FD* (100%), *psychopathology* (37%), and *factors leading to diagnosis of FD* (100%). When recording *reported occupation*, patients were only coded as 'unemployed' when this was specified by authors. Similarly, a lack of mention of patient *psychopathology* was not interpreted as an absence of comorbid psychiatric symptomatology, which was only coded when authors clearly specified that a psychiatric assessment or chart review had taken place with nothing of significance found. Marital status, race and ethnicity, and education were reported only in a small minority of cases and were therefore not addressed in this review.

*Presentation of FD* was extracted by recording the presenting sign(s), symptom(s), or diagnosis at admission. Each presentation was recorded as 'falsely reported', 'feigned', or 'induced' according to clinical information provided and categorised by medical specialty according to system affected and initial referral. Following Kanaan and Wessely (2005), where a history of repeat presentations was described, initial presentation was taken to be the index presentation for the case.

*Psychopathology* was extracted by recording current or historic psychiatric diagnoses described by the author(s). Diagnoses were not recorded where there was significant doubt expressed by the author(s) concerning the veracity of psychiatric symptoms described.

*Factors leading to diagnosis of FD* were extracted using a checklist adapted from two surveys of clinical information that might raise suspicion of FD (Bass and Halligan, 2014; Steel, 2009). This checklist included 8 items outlined with examples in Table 1. Items on the checklist were coded only if the factor contributed to the diagnosis made by the author(s). Clinical information that did not contribute to the diagnosis made by the author(s), or contributed in retrospect only, was not assessed.

## **Analysis**

IBM SPSS 23 (SPSS Inc., 2015) was used to calculate descriptive statistics. A narrative synthesis was undertaken to describe common presentations and fabrication methods reported by included studies.

## **RESULTS**

### **Demographics**

Patients with FD were described worldwide: 249 from the Americas (United States of America 237, Canada 8, Brazil 2), 150 from Europe (United Kingdom 94, Italy 9, Germany 8, Belgium 7, Greece 6, the Netherlands 6, Poland 4, Republic of Ireland 3, Austria 2, Croatia 2, Denmark 2, Spain 2, France 1, Hungary 1, Macedonia 1, Sweden 1, Romania 1), 5 from Africa (Tunisia 2, Morocco 1, South Africa 1, Zimbabwe 1), 42 from Asia (Japan 13, Turkey 13, India 6, Saudi Arabia 4, Israel 3, Iran 3), 7 from Australia, 1 from New Zealand, and 1 from Cuba.

33.8% of patients with FD were male. Mean age at presentation was 34.2 with a median of 32 years and a range of 61 (max. 79; min. 18). Table 2 contains a breakdown of age and gender by medical specialty. Patient occupation was described in 214 (47%) cases. In 122 of these cases, a healthcare/laboratory profession was reported. The single most common occupation described was nursing (N=68),

### **Index Presentation of FD**

Table 3 contains a summary of presentations of FD by medical specialty. A narrative synthesis describing common presentations and fabrication methods reported by included studies is provided in Section 3 of the Supplemental Material. Across all specialties, 22.2% falsely reported disease/injury, 19.1% simulated disease/injury, and 58.7% induced disease/injury. A full breakdown of FD severity by medical specialty is included in Table 4.

### **Psychopathology**

Presence or history of comorbid psychiatric disorders was assessed in 170 patients. The most common comorbid psychiatric disorder found in this sub-sample was depression, which was identified in 41.8% of these patients. Other common disorders that were identified included personality disorder (16.5%), substance abuse (15.3%), anxiety (14.7%), functional neurological symptoms (5.3%), and eating disorders (4.1%). 14.1% of patients reported current suicidal ideation or a history of suicide attempt(s). Authors reported the absence of comorbid psychopathology in 17.1% of the 170 cases.

## Factors leading to diagnosis of FD

In the majority of cases (78%), an *unsubstantiated presentation* contributed to a diagnosis of FD. *Past healthcare service use* contributed to diagnosis in 47% of cases, *atypical presentation* in 40% of cases, *treatment failure* in 35% of cases, *investigations indicating fabrication* in 33% of cases, *patient behaviour* in 31% of cases, *evidence of fabrication* in 31% of cases, and *information provided by patient* in 22% of cases. In two cases, the diagnosis was made solely on the basis of a spontaneous confession. A full breakdown of these factors by medical specialty may be found in Table 5.

A spreadsheet providing a basic description of all studies included in this review is provided in Section 4 of the Supplemental Material.

## DISCUSSION

### Demographic characteristics

The remarkable proportion of patients in our sample reporting an occupation related to healthcare or the laboratory (57%) supports previous observations that the majority of patients with FD claim to have worked in these settings (Ando et al., 2014; Bahna and Oldham, 2014; Baweja et al., 2014; Patenaude et al., 2006; Highland and Flume, 2002; Zamir et al., 2001; Johnson and Harrison, 2000). Krahn et al.'s (2014) chart review of 93 FD patients found that a similar proportion (44%) of patients worked in a healthcare field. Of the 122 patients who made such a claim in our review, 114 were female, supporting Krahn et al.'s (2014) identification of a subtype of FD consisting of female healthcare professionals. Over-representation of healthcare professionals in our sample may be due to publication bias, which will be discussed later. Alternatively, this result may be explained by the appeal of healthcare-related professions (in particular, nursing) for these patients. A career within a healthcare service may carry a similar appeal to deceiving clinicians for individuals predisposed to FD

(Plassman, 1994). The “rush” of undergoing medical procedures reported in FD (Lawlor and Kirakowski, 2014) may be comparable to the “rush” of delivering them in a professional setting. On balance, our findings support the broad recommendation that healthcare professionals should be particularly vigilant for FD in patients who appear to have unusual medical knowledge or claim to have worked in related occupations (Barkin et al., 2013).

The mean age of our sample at presentation (34.2 years) corroborates the results of several case series that were not included in our review (Krahn et al., 2014; Reich and Gottfried, 1983; Carney and Brown, 1983; O'Reilly and Aggeler, 1976; Petersdorf and Beeson, 1961). This finding supports the understanding held by many authors that patients with FD present to healthcare services in early adult life (Vaduganathan et al., 2014; Oner et al., 2015; Johnson and Harrison, 2000; Ameh and Speak, 2008; Donegan et al., 2012; Giuliadori et al., 2014). An exception was noted with patients presenting with dermatological problems: they tended to be older (40.6 years) and included geriatric cases (Table 2). The finding of a female majority (66.2%) in our sample may conclude an ongoing confusion in the professional literature concerning the gender distribution of patients with FD. Despite several key case series (Krahn et al., 2014; Reich and Gottfried et al., 1983; Carney et al., 1983) and reviews (Kanaan and Wessely, 2010; Eastwood and Bisson, 2008; Boyd et al., 2014; Rodriguez-Pichardo et al., 2010) indicating the opposite, numerous authors have made reference to a male predominance in cases of FD (Alicandri-Ciufelli et al., 2012; Nishimura et al., 2012) – likely a result of the inclusion of a statement to this effect in DSM-IV (American Psychiatric Association, 1994) that was subsequently reversed in DSM-IV-TR (American Psychiatric Association, 2000) and abandoned in DSM-5 (American Psychiatric Association, 2013). The case studies included in this review that were published before the release of the DSM-IV in 1994 (N=164) did not support such a statement: 58.1% of patients in this sub-sample were female. Our total sample confirms that overall, patients with FD tend to be female. However, important gender differences were observed among specialties (Table 2). For example, patients presenting with HIV-related, sexual health, or neurological problems were predominantly men, and fewer than 25% of cardiac patients were female.

## Severity of FD

Our review provided an unprecedented opportunity to compare the different methods employed by patients with FD in a large sample. We found that 58.7% of patients elected to induce illness or injury in themselves instead of attempting only to simulate (19.1%) or falsely report (22.2%) a medical problem. This preference would suggest that what has previously been regarded as an “extreme” variant of FD may in fact be its most common presentation. Patients with FD must be considered at significant risk of self-injury. This risk should be factored into any management plan for individuals suspected of medical deception. Friends, partners and family members should, if possible, be involved in this process in order to monitor the patient’s access to tools (e.g. surgical instruments) and substances (e.g. prescription drugs, poisons) that may be used to induce injury or illness. Involuntary detention may even be indicated when patients with FD are socially isolated and using methods of illness induction that are difficult to control. Abuse of insulin (to induce hypoglycaemia) and self-venesection (to induce anemia) are two such methods that were utilised by a disturbingly high number of patients in our sample and led to fatality in a number of cases. Patients with FD who adopt these methods may present themselves as medically knowledgeable, but it is unlikely that they are fully aware of the probable adverse consequences of their behaviours.

.Authors have assumed that patients with FD are intelligent and resourceful (Chambers et al., 2007; Ando et al., 2014; Highland and Flume, 2002; Barkin et al., 2013) and that these qualities are required to successfully deceive experienced clinicians (Haddad et al., 2002). However, these qualities are not required by patients with FD who induce illness or injury. Self-induction of illness or injury enables patients to reliably command attention from hospital staff (Feldman, 2008) and may reflect a need to self-harm (Lawlor and Kirakowski, 2014).

## Psychopathology

Comorbid psychiatric disorders (or their absence) were described in only 37% of cases (N=170). This finding may reflect the fact that the majority of the cases included in this review were not written by psychiatrists. Nonetheless, some observations may be made. Only 14.1% of patients were described as suicidal or as having a history of suicide attempts. It has been assumed previously that FD entails significant suicide risk and suicide attempts have been described (see Vadugnathan et al., 2014 for a useful review). In this regard, our findings may provide a degree of reassurance. Similarly, no psychotic symptomatology was described in our sample to confirm an earlier hypothesis that FD is a defence against psychosis (Ford, 1982; 1973). A surprising finding was the absence of personality disorders in all but a small minority of this sub-sample. The claim that FD is strongly associated with personality disorders (in particular, borderline personality disorder) is widespread in the professional literature (Oner et al., 2015; Baweja et al., 2014; Vadugnathan et al., 2014; Gordon and Sansone, 2013; Lin et al., 2012; Schulz and Strauch, 2008; Patenaude et al., 2006) and has been included in multiple review articles (Steel, 2009; Bass and Halligan, 2014). Even so, the comorbid diagnosis reported most commonly in our sample was depression, providing support instead for an association between FD and mood disturbance (Bass and Halligan, 2014; Pascual et al., 2001). However, the relationship between these two diagnoses is not clear. FD may be truly comorbid with depression due to shared risk factors for the two disorders, which include childhood abuse or neglect (Norman et al., 2012; Chen et al., 2010), parental failures (Otowa et al., 2013; Gao et al., 2012; Sakado et al., 2000; Kendler et al., 2000), marital difficulties (Whisman et al., 2000), substance abuse (Bovasso et al., 2014; Boden et al., 2011; Conner et al., 2008), and stressful life events (Stroud et al., 2008; Hammen, 2005). Alternatively, FD may be secondary to depression – for example, as an expression of low self-esteem, or a manifestation of the urge to self-harm, which has been linked to depressive symptoms (Haw et al., 2001; Briere and Gil, 1998). In this case, it is plausible that treatment of depressive symptoms in cases of FD may lead to a reduction of factitious illness behaviour.



## Presentation of FD

Considerable variation was observed in the number of cases included in this review for each medical specialty. This variation may be explained by differential interest in FD among authors working in different specialties (Kanaan and Wessely, 2010). For example, the high number of dermatology cases eligible for this review may be the result of dermatologists' increased awareness of or interest in factitious disorder rather than a genuine "preference" of patients with FD. The inclusion of 'dermatitis artefacta' in the ICD predates the inclusion of FD by several decades (World Health Organisation, 1975; 1948). Alternatively, variation among specialties may be explained by the relative difficulty of identifying FD within various medical specialties. For example, the preponderance of factitious hypoglycaemia in this review may signal the comparative ease with which insulin abuse can be detected in the laboratory (Kinns et al., 2013; Kenedi et al., 2011). However, assuming that the distribution of cases across medical specialties corresponds to some degree to preferences of patients with FD, our findings demonstrate the need for health professionals working in endocrinology, cardiology, and dermatology services to be specially watchful for FD.

As expected, patients with FD gravitated towards signs and symptoms leading to protocol-driven or "fast-track" admission, such as retrosternal chest pain. Similarly, patients made good use of widely available agents to induce serious illness, such as insulin, anti-coagulants, or thyroid hormones. This may have contributed to the high number of patients presenting with dermatological and endocrinological problems. It is clear that patients with FD are capable of using their medical knowledge not only to simulate illness convincingly, but also to find the "path of least resistance" to admission. Nevertheless, many patients were attracted to specialties with more complex disorders and a greater likelihood of discovery, such as cardiology and neurology. Kanaan and Wessely (2005) discussed this problem in their own review of neurological cases of FD, suggesting that the increased difficulty of simulating certain medical problems (due to modern imaging and laboratory investigations) may in FD be counterbalanced by increased 'reward' in the form of greater attention

and sympathy. This suggestion would explain the relative popularity of oncology and cardiology to patients in our sample, despite the difficulty often involved in fabricating cancer or coronary disease.

However, we might suggest that it is simply the case that patients with FD are able to make as much use of modern technology to maintain their deception as clinicians are to detect it, and for this reason it is not as difficult for these patients to fabricate complex medical problems as we might assume. Numerous authors included in this review discuss the ease with which their patients were able to use the Internet to research their presentation of choice (Hariharasubramony et al., 2012; King et al., 2008), forge medical reports or referral letters (Griffiths et al., 2009; Levenson et al., 2007), and even purchase prescription medications (Saiyasombat et al., 2012). The Internet may therefore enable patients with FD to be sufficiently versatile and adaptive in their deception to present to more challenging medical specialists (Ackermann et al., 2000). In any case, our findings challenge the notion that the problems presented by FD will be overcome with innovations in health technology.

### **Factors leading to diagnosis of FD**

As may be expected, a presentation unsubstantiated by objective clinical evidence was found to have facilitated discovery of FD in the majority of cases included in this review across all specialties. Our review therefore supports existing guidelines that caution health professionals to consider FD early when encountering patients whose complaints appear unsupported by physical examination, the results of investigations, and so forth (Bass and Halligan, 2014; Steel, 2009). However, the variance among medical specialties that we observed in clinicians' use of other sources of information to diagnose FD is unaccounted for by these guidelines, and contrasts sharply with several claims made in the published literature. For example, it has been assumed that dermatologists are able to diagnose FD primarily on the basis of physical examination, because skin lesions produced by patients with FD are morphologically atypical (Wojewoda et al., 2012; Fujiwara et al., 2008; Livaoglu et al., 2008) and located on easily accessible areas of the body (Gregurek-Novak et al., 2005). While this clinical picture facilitated diagnosis in 70% of the dermatological cases of FD that we reviewed, in a

significant minority of cases the appearance of lesions did not alert dermatologists to the possibility that they were being deceived. Similarly, although it has been argued that patients presenting with factitious endocrinological illness may be recognised by their surgical history (Thynne et al., 2014), in the majority of cases involving such patients, such a history was either not described or not regarded as suspicious by endocrinologists. Future guidelines for the detection of FD in clinical settings must be data-driven and sensitive to the practical realities of diagnosing the disorder in different medical specialties. This approach will help to avoid problematic assumptions of the kind discussed above, and offset the limitations of “one size fits all” clinical guidelines for FD. Our findings, accrued from the largest sample of patients with FD analysed to date, and a broad range of medical specialties, provide a robust starting-point for such an approach.

### **Limitations**

A number of limitations to this review must be acknowledged. Firstly, although case reports constitute the best source of knowledge currently available about FD, the sample used in this review is non-random and unlikely to be fully representative. Studies appeared more likely to be published if they presented an unusual manifestation of FD, a novel technique for detecting FD, or an entertaining account. A culture of “one-upmanship” may therefore have deterred potential authors from submitting cases of FD that were less severe, or similar to previously published cases. Publication bias of this kind may also have accounted for the over-representation of healthcare professionals in our sample. Healthcare professionals are clearly capable of using their expertise to fabricate medical need more convincingly, as is shown in several reports included in this review (Bahna et al., 2014; Norcliffe-Kaufmann et al., 2010; Farrier and Mansel, 2002). It therefore stands to reason that cases involving healthcare professionals would be over-represented in a literature biased towards ingenious cases. This publication bias may be compared to the ‘file-drawer problem’ described in meta-analysis (Rotton et al., 1995), although the preponderance of less severe and non-novel cases of FD included in our sample provides reassurance.

Secondly, it is plausible that our sample contains duplicates, as patients with FD are typically treated by several clinicians in the course of their deception - potentially across several regions (Maur et al., 1973) or countries (Addison and Talan, 1974) - any of whom may publish the case (Patenaude et al., 2006). Cases explicitly describing a previously reported patient were excluded, but publication of cases in low-impact journals with a limited readership may have restricted the extent to which authors could be aware that they were describing the same patient as a previous case report.

Thirdly, because we extracted only basic demographic and clinical information from our sample, we could not examine the relationship between the results of this review and patient outcomes - nor could we relate our data to the aetiology of FD. Eastwood and Bisson (2008) conducted a similar systematic review of cases ( $N = 316$ ) in order to evaluate management techniques for FD. However, inconsistent reporting of outcomes in case studies of FD significantly limited the extent to which they were able to use their analysis to make recommendations for the treatment of the disorder. This would suggest that original research is required to address these research questions assertively.

Finally, although care was taken to include only cases meeting a highly conservative interpretation of the DSM-5 criteria for FD (Krahn et al., 2014), it is possible that factors integral to the diagnosis of FD (e.g., absence of external reward) were not considered by authors, as many healthcare professionals do not encounter FD in everyday clinical practice and are unfamiliar with its diagnostic criteria (Koufagued et al., 2015). Indeed, the most convincing patients with FD do not appear to meet diagnostic criteria for the disorder at all. Cases of FD reported in the professional literature (and consequently, in this review) may represent only the accounts of the patients least capable of avoiding detection, or the clinicians most capable of detecting them.

## Conclusion

FD is one of the most challenging disorders in medical experience but the clinical and demographic profile of the disorder has yet to be clarified with a sufficiently large sample.. Accordingly, we conducted a systematic review of 455 cases of FD in the professional literature, the largest sample analysed to date. Our findings provide several clinical recommendations (see below) and a strong first step towards an evidence-based approach to detection and treatment of FD..

### Clinical recommendations

- Clinicians should be particularly vigilant for FD in patients who are female, in early adult life, and claiming to have worked in healthcare or a laboratory
- Although patients with FD may appear in any specialist setting, endocrinology, cardiology and dermatology services should expect to encounter more
- FD is associated with low suicide risk, but these patients typically self-induce illness or injury and should therefore be considered at high risk of permanent damage, if not fatality
- FD is associated with depressive symptoms more than personality disorders, and may be improved by treatments for depression

**Table 1. Factors leading to diagnosis of FD**

#	Factor	Examples
1	Past healthcare service use	History of extensive healthcare service use; history of peregrination between healthcare services; history of FD confirmed by healthcare professional
2	Information provided by patient	Inconsistent, selective or misleading biographical information provided; evasive when history is taken; dramatic but unlikely medical history provided; unusual difficulty corroborating information provided; refusal to allow access to outside information sources
3	Atypical presentation	Symptoms predominantly occur when the patient is not under observation; course of illness is impossible, highly improbable, or does not follow the natural history of the presumed diagnosis
4	Unsubstantiated presentation	Investigations normal or inconclusive
5	Evidence of fabrication	Physical evidence of fabrication discovered through search or surveillance; patient directly witnessed simulating disease
6	Patient behaviour	Unusual medical knowledge or use of medical terminology; eagerness for medical procedures; aggression or defensiveness with healthcare staff; non-compliance with diagnostic or treatment recommendations; <i>pseudologia fantastica</i> ; patient opposes psychiatric involvement while pursuing medical or surgical options
7	Investigations indicating fabrication	Investigations reveal mechanism of fabrication; investigations rule out organic aetiology; evidence from investigations contradicts information provided by patient
8	Treatment failure	Appearance of new symptoms on commencement of treatment; symptoms worsen on commencement of treatment

**Table 2. Basic demographic characteristics of patients diagnosed with FD by medical specialty**

Specialty	N	Age (SD; range [min; max])	% Female
Allergy & Immunology	8	27.9 (10.3; 31 [min 20; max 51])	88
Cardiology	44	38.4 (11.9; 47 [min 20; max 67])	23
Dermatology	43	40.6 (16; 61 [min 18; max 79])	79
Endocrinology	59	32.3 (9.4; 36 [min 18; max 54])	78
ENT	11	28.3 (10.9; 37 [min 19; max 56])	73
Gastroenterology	29	34.1 (11; 43 [min 19; max 62])	76
Haematology	27	34.4 (14.4; 53 [min 21; max 74])	74
HIV & Sexual Health	11	32 (12; 43 [min 19; max 62])	45
Microbiology & Infection	13	28.5 (6.3; 25 [min 18; max 43])	92
Neurology	32	34.4 (10; 46 [min 22; max 68])	44
Obstetrics & Gynaecology	7	40.3 (16.1; 47 [min 21; max 68])	100
Oncology	12	31.6 (7.9; 26 [min 19; max 45])	92
Ophthalmology	18	32.1 (15.2; 55 [min 18; max 73])	83
Oral & Maxillofacial	8	27.4 (5.3; 15 [min 20; max 35])	75
Orthopaedics & Trauma	34	30.1 (9.9; 38 [min 18; max 56])	53
Plastic & Reconstructive Surgery	13	33.4 (9.4; 31 [min 23; max 54])	77
Pulmonary & Respiratory	33	33 (12.8; 53 [min 19; max 72])	70
Rheumatology	9	36.9 (8.2; 25 [min 22; max 47])	67
Urology & Nephrology	30	34.9 (9.9; 35 [min 22; max 57])	53
Other	14	41.7 (8.5; 31 [min 22; max 53])	71

**Table 3. Index presentations of cases by medical specialty**

Specialty	N	Index presentations
Allergy & Immunology	8	Allergic emergency induced (N=3), signs of immune deficiency induced (N=3), allergic emergency simulated (N=2)
Cardiology	44	Retrosternal chest pain reported (N = 29), hypertension induced (N = 5), arrhythmia induced (N = 2), unconsciousness and bradycardia induced (N = 1), history of myocardial infarction reported (N = 1), syncopal episodes simulated (N = 3), syncopal episodes induced (N = 1), history of syncopal episodes reported (N = 1), ventricular tachycardia and myocardial infarction induced (N = 1)
Dermatology	43	Generalised lesions induced (N = 10), breast lesions induced (N = 7), facial lesions induced (N = 4), leg lesions induced (N = 3), pyoderma gangrenosum simulated (N = 3), pyoderma gangrenosum induced (N = 1), hand lesions induced (N = 2), genital lesions induced (N = 2), purpura of the knee induced (N = 2), subcutaneous nodules and abscesses induced (N = 2), ulceration of elbow and forearm induced (N = 1), acute erythematous eruption induced (N = 2), neck lesions induced (N = 1), arm lesions induced (N = 1), generalised purulent lesions with lichenification induced (N = 1), swelling of the hand induced (N = 1), painful lobules of lower body induced (N = 1), cheilorrhagia and cheilitis of lip induced (N = 1)
Endocrinology	59	Recurrent hypoglycaemia induced (N = 31), signs and symptoms of Cushing's syndrome induced (N = 9), thyrotoxicosis induced (N = 8), diabetic ketoacidosis induced (N = 4), history of diabetes reported (N = 2), hyperglycemia induced (N = 1), pheochromocytoma simulated (N = 1), endocrine neoplasia simulated (N = 1), symptoms and family history of multiple endocrine neoplasia reported (N = 1), history of Zollinger-Ellison syndrome reported (N = 1)
ENT	11	Facial swelling induced (N = 4), airway distress reported (N = 2), bleeding from mouth, nose, ears and eyes simulated (N = 1), bleeding from ears simulated (N = 1), airway distress feigned (N = 1), cheilitis of lip induced (N = 1), ear drainage simulated (N = 1)
Gastroenterology	29	Severe diarrhoea induced (N = 6), diarrhoea simulated (N = 2), recurrent vomiting induced (N = 1), Haematemesis simulated (N = 5), haematemesis and haematochezia reported (N = 1), haematemesis induced (N = 1), gastrointestinal bleeding induced (N = 3), epigastric pain induced (N = 3), rectal bleeding simulated (N = 1), gastrointestinal bleeding simulated (N = 1), deterioration of Crohn's disease induced (N = 2), uro-intestinal fistulae simulated (N = 1)
Haematology	27	Anemia induced (N = 9), purpura induced (N = 4), hypercalcemia induced (N = 2), hypokalemia induced (N = 1), systemic mastocytosis reported (N = 1), acute lymphoblastic leukemia reported (N = 1), chronic myeloid leukemia reported (N = 1), haemophilia reported (N = 1), epistaxis induced (N = 1), abnormal coagulation induced (N = 1), signs of deep vein thrombosis simulated (N = 2), sickle cell disease reported (N = 3)
HIV & Sexual Health	11	History of HIV reported (N = 5), history of AIDS reported (N = 3), history of HIV-related Kaposi's sarcoma reported (N = 1), history of AIDS-related disease reported (N = 1), history of venereal disease reported (N = 1)
Microbiology & Infection	13	Sepsis induced (N = 7), septic arthritis induced (N = 3), necrotising fasciitis simulated (N = 1)



Neurology	32	Chronic pain reported (N = 7), paralysis or weakness simulated (N = 4), unconsciousness simulated (N = 4), cyclic hypersomnia simulated (N = 1), seizures simulated (N = 3), torsion dystonia simulated (N = 2), hemifacial spasm simulated (N = 1), symptoms of acute meningitis reported (N = 2), migraine reported (N = 1), scalp abrasions induced (N = 2), signs of baroreflex failure induced (N = 1), sciatica and urinary incontinence reported (N = 1), blindness simulated (N = 1), aphasia simulated (N = 1), deterioration of Parkinson's induced (N = 1)
Obstetrics & Gynaecology	7	Vaginal bleeding induced (N = 5), menorrhagia induced (N = 1), vaginal discharge simulated (N = 1)
Oncology	12	History of breast cancer reported (N = 4), family history of breast cancer reported (N = 2), ovarian cancer reported (N = 1), cancer of small intestine reported (N = 1), uterine cancer reported (N = 1), Hodgkin's disease reported (N = 1), adenocarcinoma of urinary bladder reported (N = 1), symptoms of osteogenic carcinoma reported (N = 1)
Ophthalmology	18	Keratoconjunctivitis induced (N = 6), corneal damage induced (N = 4), anterior scleritis induced (N = 2), diplopia reported (N = 2), acute endophthalmitis induced (N = 1), eyelid swelling induced (N = 1), crystalline keratopathy induced (N = 1), signs of basal cell carcinoma induced (N = 1)
Oral & Maxillofacial	8	Swelling of mandibular region induced (N = 2), abrasion of oral mucosa induced (N = 2), gingival ulceration induced (N = 1), progressive facial pain reported (N = 1), subluxation of jaw simulated (N = 1)
Orthopaedics & Trauma	34	Subcutaneous emphysema induced (N = 5), chronic wound deterioration induced (N = 5), pain induced (N = 5), severe trauma simulated (N = 4), severe trauma induced (N = 3), subfascial emphysema induced (N = 2), joint dislocation simulated (N = 2), joint dislocation induced (N = 1), chronic edema induced (N = 1), burns induced (N = 1), pyoderma gangrenosum induced (N = 1), trauma reported (N = 1), thigh abscess induced (N = 1), suprapubic ulceration and vesicocutaneous fistula induced (N = 1)
Plastic & Reconstructive Surgery	13	Wound deterioration induced following surgery (N = 9), skin ulceration induced (N = 3), deep muscular abscess induced (N = 1)
Pulmonary & Respiratory	33	Asthmatic episodes simulated (N = 8), acute respiratory distress simulated (N = 3), haemoptysis simulated (N = 4), haemoptysis reported (N = 3), cystic fibrosis reported (N = 2), pleuritic chest pain reported (N = 2), intractable bronchorrhea reported (N = 1), severe leg pain and pulmonary history reported (N = 1), asphyxia simulated (N = 1), inability to be weaned from ventilator reported (N = 1), pneumothorax induced (N = 2), hypoxemia induced (N = 1), signs of collagen vascular disorder induced (N = 1), inhalational pulmonary talcosis induced (N = 1)
Rheumatology	9	Lobular panniculitis induced (N = 2), nodular panniculitis induced (N = 1), non-specific panniculitis induced (N = 2), polyarthralgia and subcutaneous masses induced (N = 1), systemic lupus erythematosus reported (N = 2), arthritis simulated (N = 1),
Urology & Nephrology	30	Severe renal pain reported (N = 13), UTI induced (N = 5), haematuria simulated (N = 3), proteinuria simulated (N = 1),

**Table 4. FD severity by medical specialty**

<b>Specialty</b>	<b>N</b>	<i>False report of disease/injury</i>	<i>Feigned disease/injury</i>	<i>Induced disease/injury</i>
Allergy & Immunology	8	0%	25%	75%
Cardiology	44	61%	16%	23%
Dermatology	43	0%	0%	100%
Endocrinology	59	7%	2%	92%
ENT	11	9%	45%	45%
Gastroenterology	29	10%	17%	72%
Haematology	27	26%	0%	74%
HIV & Sexual Health	11	100%	0%	0%
Microbiology & Infection	13	0%	0%	100%
Neurology	32	31%	56%	13%
Obstetrics & Gynaecology	7	0%	29%	71%
Oncology	12	92%	0%	8%
Ophthalmology	18	6%	6%	89%
Oral & Maxillofacial	8	38%	0%	63%
Orthopaedics & Trauma	34	3%	15%	82%
Plastic & Reconstructive Surgery	13	0%	0%	100%
Pulmonary & Respiratory	33	27%	58%	15%
Rheumatology	9	11%	22%	67%
Urology & Nephrology	30	27%	50%	23%
Other	14	29%	36%	36%
<b>Median</b>		<b>10.5%</b>	<b>15.5%</b>	<b>69%</b>
<b>IQR</b>		<b>29.25</b>	<b>34.25</b>	<b>64.25</b>

**Table 5. Factors leading to diagnosis of FD by medical specialty**

<b>Specialty</b>	<b>N</b>	<i>Past healthcare service use</i>	<i>Information provided by patient</i>	<i>Atypical presentation</i>	<i>Unsubstantiated presentation</i>	<i>Evidence of fabrication</i>	<i>Patient behaviour</i>	<i>Investigations indicating fabrication</i>	<i>Treatment failure</i>
Allergy & Immunology	8	25%	0%	50%	63%	50%	38%	38%	50%
Cardiology	44	68%	41%	27%	91%	23%	50%	21%	32%
Dermatology	43	23%	5%	70%	79%	26%	16%	30%	42%
Endocrinology	59	29%	12%	31%	78%	32%	10%	73%	36%
ENT	11	73%	27%	82%	91%	55%	73%	18%	27%
Gastroenterology	29	38%	14%	35%	86%	41%	24%	52%	28%
Haematology	27	70%	22%	30%	78%	41%	22%	41%	19%
HIV & Sexual Health	11	82%	46%	18%	91%	9%	46%	0%	18%
Microbiology & Infection	13	54%	8%	15%	54%	69%	23%	62%	54%
Neurology	32	59%	28%	66%	84%	13%	63%	9%	31%
Obstetrics & Gynaecology	7	57%	0%	29%	71%	29%	14%	43%	43%
Oncology	12	67%	75%	25%	50%	17%	25%	0%	0%
Ophthalmology	18	44%	6%	50%	67%	28%	44%	28%	33%
Oral & Maxillofacial	8	50%	0%	38%	75%	63%	25%	0%	100%
Orthopaedics & Trauma	34	44%	35%	35%	56%	24%	27%	32%	32%
Plastic & Reconstructive Surgery	13	31%	15%	46%	69%	23%	31%	39%	77%
Pulmonary & Respiratory	33	55%	24%	46%	88%	27%	27%	15%	49%
Rheumatology	9	44%	22%	67%	89%	56%	33%	11%	44%
Urology & Nephrology	30	33%	20%	10%	90%	43%	23%	23%	17%
Other	14	50%	36%	43%	64%	7%	43%	36%	29%
<b>Median</b>		<b>50</b>	<b>21</b>	<b>36.5</b>	<b>78</b>	<b>28.5</b>	<b>27</b>	<b>29</b>	<b>32.5</b>
<b>IQR</b>		<b>30.75</b>	<b>26.75</b>	<b>22.5</b>	<b>24</b>	<b>25.25</b>	<b>20.75</b>	<b>28.5</b>	<b>20.5</b>

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